Squamous Cell Carcinoma in a Transgender Man’s Neo-Urethra after Metoidioplasty

Maria J. D’Amico, Eric Kirshenbaum, Chris M. Gonzalez

Boston University School of Medicine, Boston, MA; Department of Urology, Loyola University Medical Center, Maywood, IL, USA

Abstract

Metoidioplasty is a gender-affirming surgery for transgender men, which creates a neophallus using vaginal mucosa and labia majora. One known complication of this procedure is urethral stricture. We report the novel case of a patient with urethral stricture 4 years after metoidioplasty found to have squamous cell carcinoma in situ likely originating from the vaginal mucosa grafted to the neourethra, and highlight the importance of sending such strictures for pathologic evaluation.

Case Report

The patient is a 39-year-old transgender male who underwent a metoidioplasty with total vaginectomy and urethral lengthening at an outside hospital. His postoperative course was uneventful until he began experiencing dysuria and urinary frequency 2 years following the metoidioplasty. At the time, he underwent cystoscopy, which was reported to be normal. Three years following his initial surgery, he had a weakened stream, and eventually presented to our emergency department in urinary retention. Urology was consulted and successfully placed a 12Fr silicone catheter.

He was subsequently taken to the operating room for cystoscopy and retrograde urethrogram, which revealed narrowing of the meatus with stenosis of the distal 2/3 of the neourethra at the junction between the native urethra and pars fixa. He was later taken to the operating room for definitive repair. Upon exploration, he was noted to have a 4–6Fr urethral stenosis with significant scarring throughout. The non-viable scar was excised and sent to pathology for analysis. Given the degree of scarring, a first stage Johanson urethroplasty with a perineal urethrostomy to the native urethra was completed. Endoscopy of the native urethra and bladder was unremarkable. His immediate postoperative course was unremarkable.

Post-operatively the pathology results from the urethral stricture were reported as squamous cell carcinoma (SCC) in situ/high
grade squamous intraepithelial lesion, p16 positive, and negative for invasive carcinoma. Three weeks after the procedure, the patient had recovered well and was voiding without issue. Examination revealed no mucosal or skin abnormalities. He is to follow-up for surveillance cystoscopy and exam under anesthesia in 2 months.

Discussion

Our case was unique in that it is the first reported instance of SCC in situ in a neo-urethra after a metoidioplasty. Notably, the number of transgender patients undergoing gender affirmation surgery is increasing. Of all transgender patients, 10–30% will seek gender-affirming surgery [4]. Transgender male patients who desire gender affirming surgery are treated with phalloplasty or metoidioplasty.

The patient in this case underwent a metoidioplasty with total vaginectomy. Goals of the procedure are to achieve voiding in the standing position via creation of an esthetically pleasing masculine genitalia, along with preservation of clitoral sensation. A hormonally enlarged clitoris is used to create the neophallus, and the labia majora is used to create a scrotum. A neourethra consisting of tubularized labia minora is anastomosed to the native urethra. Buccal mucosa may be utilized for further urethral reconstruction and creation of a new urethral plate [1].

Complications after metoidioplasty include minor issues such as dribbling and spraying with urination, as well as more complex complications such as urethral stricture or fistula. Nikolavsky et al. [3] described urethrocutaneous fistula as the most common complication after metoidioplasty or phalloplasty, often arising at an anastomotic site along the urethra. Urethral strictures also commonly occur at anastomotic sites, likely due to poor perfusion and resultant ischemia, as well as urine leaks contributing to scar formation. Urethral strictures must be promptly recognized and treated, as they can lead to obstruction and fistulae with eventual infection or sepsis.

These surgeries are performed at few institutions worldwide. Dy et al. [5] identified patients who underwent revisional surgery for complications of masculinizing gender-affirming surgery at outside institutions. Of the 55 patients, a majority of patients had a stricture or meatal stenosis (86%). The strictures were repaired using a variety of methods including direct vision internal urethrotomy, perineal urethrostomy, stricturoplasty, anastomotic urethroplasty, buccal mucosa graft urethroplasty, rotational fasciocutaneous flaps, and various meatoplasty techniques. The wide variety of treatment highlights the lack of standardized approaches to address these complications. This study notes the importance of education of reconstructive urologists to prepare them to evaluate and treat patients with complex surgical needs who may have been treated elsewhere. For our patient, creation of a perineal urethrostomy was used because the stricture was along the entirety of the metoidioplasty. We would like to highlight the importance of sending the excised tissue to pathology for analysis and p16 testing, as the SCC in situ would not have been discovered otherwise.

Our patient presumably had SCC of the vaginal mucosa which was grafted to form an extension of the native urethra. Vulvar SCC is often associated with high risk human papilloma-virus (HPV), particularly HPV 16 subtype, which tends to occur in younger individuals such as our patient [6]. The gold standard for diagnosis of vulvar SCC is a biopsy for histopathological examination. Treatment for SCC in situ includes topical imiquimod therapy, 5-fluorouracil, ablative laser therapy, surgery, or photodynamic therapy with Mohs surgery, which achieves > 90% complete response rate [7]. In our patient, we hypothesize that the resultant malignancy may have been due to undiagnosed HPV along with irritation from anastomosis and resultant stricture.

Conclusion

Metoidioplasty involves urethral extension using tissue that may be exposed to HPV. This puts the neourethra at risk for SCC, especially in the setting of chronic inflammation associated with urethral strictures. While SCC of the neourethra after metoidioplasty is rare, it may lead to complications requiring revision of the gender-affirming surgery. It is important for physicians to include SCC in the differential diagnosis of complications of gender affirming surgery and always send tissue for histopathological evaluation and HPV testing at the time of revision.
References